<Case Report>Leiomyosarcoma of the Colon in a Newborn : a Case Report and Review of the Literature

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CITATION:

IKEDA, SHINJI ...[et al]. <Case Report>Leiomyosarcoma of the Colon in a Newborn : a Case Report and Review of the Literature. 日本外科宝函 1993, 62(3): 166-171

ISSUE DATE:

1993-05-01

URL:

http://hdl.handle.net/2433/203678

Arch Jpn Chir 62(3), 166～171, Mai., 1993

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Leiomyosarcoma of the Colon in a Newborn : a Case Report and Review of the Literature

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Received for Publication, Feb. 5, 1993

Abstract

A rare case of leiomyosarcoma of the ascending colon in a 26-day-old boy, presenting with tarry stools and an abdominal mass, is reported, and the literature pertaining to childhood colorectal leiomyosarcoma is reviewed.

Introduction

Leiomyosarcoma is the most common non-epithelial malignancy. In adult, it is most common in the stomach, followed by the small bowel, and least common in the colon and rectum[1]. The tumor is extremely rare in children. We believe that this is the sixteenth reported case of colorectal leiomyosarcoma in the world literature.

Case Report

A 26-day-old boy was transferred to our surgical ward because of tarry stools. A small egg-shaped mass was palpable in the right lower quadrant of the abdomen. Barium enema revealed extreme narrowing of the ascending colon (Fig. 1). At surgery, a tumor originating from the wall of the ascending colon was found (Fig. 2). No intra-abdominal metastases were found. A right hemicolectomy with ileo-transverse anastomosis was performed and the baby recovered well. The sessile tumor, which almost filled the lumen of the bowel, was a well-defined mass. There was superficial ulceration. On cut section, the mass showed a glistening white tumor with areas of necrosis. The histologic diagnosis was leiomyosarcoma. The patient received no further treatment and has remained well since then 6 years.

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Key words: Leiomyosarcoma, Colon, Children
索引用語: 平滑筋肉腫，大腸，小児

Pathology

The tumor, measuring 4.7 × 4.5 × 2.1 cm in diameter, was situated in the submucosa, and ex-

Fig. 1 Barium enema demonstrating extreme narrowing of the ascending colon

Fig. 2 The tumor originating from the wall of the ascending colon

tended to the serosa. The tumor was highly cellular and was composed of slender or slightly plump cells arranged in fascicles of varying sizes. A pericytoma-like pattern was another feature. The cytoplasm was usually eosinophilic, with some clear cells. Occasional mitotic figures were detected (Fig. 3). The features are compatible with intestinal leiomyosarcoma.

Discussion

Leiomyosarcoma of the colon and rectum in children are rare; only fifteen cases have been reported (Table 1). The first case of leiomyosarcoma of the colon in an infant was recorded by LONGINO in 1958. The ages of the cases published in the literature vary from a few days of life to 15 years of age, with a female to male ratio of 2,2 : 1. This female predominance is the opposite of that reported in adults[1]. The predominant sings of leiomyosarcoma of the colon in children appear to be obstruction, palpable masses, and perforation. Four of the fifteen cases had obstruction, four perforation, and three had a mass. There has been controversy about the criteria for the diagnosis of leiomyosarcoma. ALTMAN[2] and WURLITZERS[17] criteria are based on the number of mitosis, anaplasia, and invasion of surrounding structures. AKWARI et al[1] proposed that even if one mitotic figure per 10 high-power fields is found, the tumor should be considered malignant. In our patient, some mitoses existed and there was invasion of the surrounding structures. Therefore, the tumor was diagnosed as malignant. Radical resection was the treatment of choice, since leiomyosarcoma is considered to be resistant to chemotherapy and radiation[1,3]. ANGERPOINTNER[4] and MARQUEZ[11], however, used adjuvant chemotherapy, and SZTANKAY[15] used both surgery and radiation in their-reported cases.

In adults, the prognosis of leiomyosarcoma is not good[14], but all of the pediatric patients except

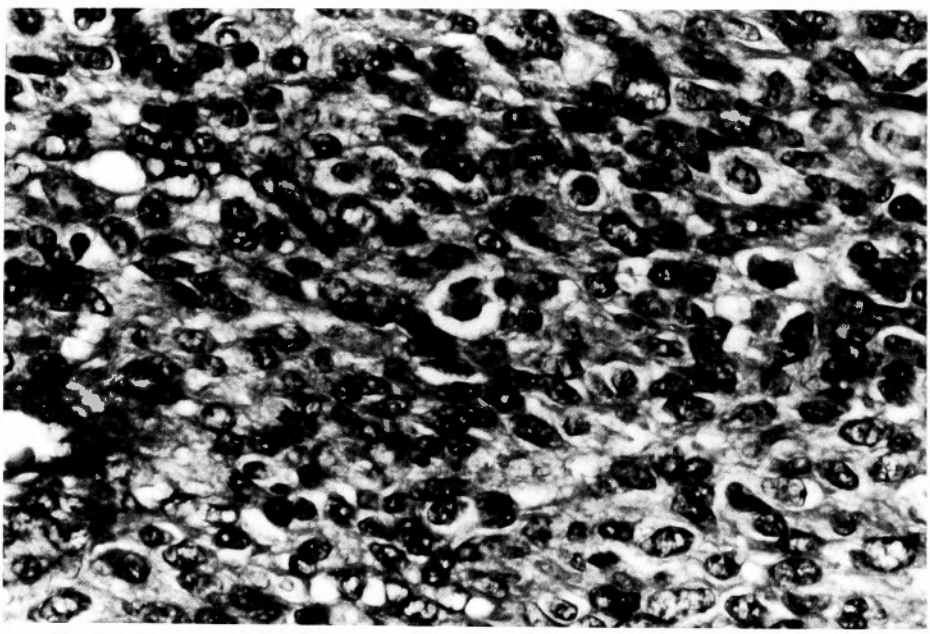

Fig. 3 Histologically, the tumor displays the spindleshaped cells and mitotic figures characteristic of leiomyosarcoma, (H&E, original magnification ×200)

Table 1 Reported Cases of Childhood Colorectal Leiomyosarcoma

Author	Year	Age at diagnosis	Sex	Presenting feature	Site	Treatment	Follow up	Additional therapy
Longino[2]	1958	Infant	F	Mass	Transverse colon	Resection	Well 1 yr	
Sztankay[3]	1959	10 yr	F	Bloody stool	Rectum	Resection	Well 5 yr	Radiation
Kriss[4]	1960	4 wk	F	Obstruction	Transverse colon	Resection	Well 1 yr	
Valentini[5]	1961	12 yr	F	Pyrexia	Ascending colon	Hemicolectomy	No	
Hicsonmey[6]	1975	12 dys	F	Mass protruding Through anus	Rectum	Resection	Well 7 mo	
Ein[7]	1979	3 dys	M	Obstruction	Transverse colon	Resection	Well 13 yr	
Ein[7]	1979	Newborn	M	Perforation	Transverse colon	Resection	Well 6 yr	
Fischer[8]	1980	Newborn	M	Perforation	Descending colon	Hemicolectomy	Well 5 yr	
Angerpointner[9]	1981	4 yr	M	Mass	Ascending colon	Hemicolectomy	Well 18 mo	Chemotherapy
Posen[10]	1981	7 wk	F	Obstruction Perforation	Sigmoid colon	Resection	Died postop	
Ayoub[11]	1986	15 yr	F	Mass	Sigmoid colon	Resection	Died 9 mo	
Marquez[12]	1986	8 mo	F	Obstruction	Transverse colon	Resection	Well 4 yr	Chemotherapy
Nagaya[13]	1987	Newborn	F	Perforation	Transverse colon	Resection	Well 17 mo	
Delucchi[14]	1988	5 yr	F	Abdominal pain	Sigmoid colon	Resection	Well 3 yr	
Angel[15]	1992	5 yr	F	?	Sigmoid colon	Resection	Well 3 yr	
Current Case	1992	Newborn	M	Mass, bloody stool	Ascending colon	Hemicolectomy	Well 6 yr	

two were alive at the time they were reported. This may reflect their early discovery, or may indicate a different biological nature of this disease in children.

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和文抄録

新生児の上行結腸平滑筋肉腫一例報告と文献的考察

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生後26日目の男児，下血，腹部腫瘤を主訴に当科紹介受診．注腸造影，CT スキャン等にて上行結腸腫瘍を疑い，開腹術施行．腫瘍は上行結腸より発生しているのが認められ，右半結腸切除術を行った．腫瘍の大きさ 4.7×4.5×2.1 cm，病理組織学的検査にて平滑筋肉腫と診断された．大腸の平滑筋肉腫は小児には極めてまれであり，今までに15例の報告をみるにすぎない．今回，文献的考察を加えて報告した．